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Defining and measuring recovery from myalgic encephalomyelitis and chronic fatigue syndrome: The physician perspective

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Abstract

Purpose: To inform an operationalized definition of recovery from myalgic encephalomyelitis and chronic fatigue syndrome for research and practice. Without a consensus on defining and measuring recovery, there will continue to be controversy amongst researchers, clinicians, and patients when interpreting treatment outcomes.

Method: This study explores physicians' views on recovery from myalgic encephalomyelitis and chronic fatigue syndrome. We conducted semi-structured interviews with 10 physician participants who are experts in the myalgic encephalomyelitis and chronic fatigue syndrome field. Our deductive thematic analysis, using a realist perspective, provided a framework for differentiating recovery and significant improvement.

Results: Physicians conceptualized recovery as complete symptom remission and a return to premorbid functioning (adjusted for with age), whereas they viewed significant improvement as a substantial reduction in symptoms with considerable functional gains, where patients may operate in daily life but still must cope or be treated.

Conclusions: Our findings provide recommendations and approaches for measuring: daily functioning, symptomatology, quality of life, and physical functioning.

Keywords

chronic illness; assessment; prognosis; physical functioning; psychosocial functioning; qualitative

Myalgic encephalomyelitis (ME) and chronic fatigue syndrome (CFS) have detrimental effects on patients' social, occupational, and leisurely lives. Although evidence has demonstrated differences between ME and CFS [1], these illnesses share many symptoms and are often studied together. Numerous symptoms characterize ME and CFS, including post-exertional exacerbations (commonly referred to as post-exertional malaise; PEM), impairment of memory and concentration, unrefreshing sleep, arthralgia and/or myalgia, and

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several autonomic, neuroendocrine, and immune manifestations [2,3,4]. While ME and CFS are chronic and often debilitating [4], few studies have examined recovery from these illnesses.

There are currently no operationalized criteria for recovery from ME and CFS [for review, see 5]. This lack of a definition is problematic for patients, researchers, and healthcare providers. Reviewing 14 treatment studies, Cairns and Hotopf [6] found a median recovery rate of 5% for ME and CFS. However, criteria for both recovery and improvement have been inconsistently defined across studies, making it difficult to compare and assess treatment outcomes. As an example, in their cognitive behavioral therapy (CBT) intervention, Knoop, Bleijenberg, Gielissen, van der Meer, and White [7] reported recovery from a single domain of CFS (e.g., no physical disability) and recovery from multiple domains (e.g., fatigue, health perceptions). Around 59–66% of patients recovered when considering single domains, but only 23% recovered when considering multiple domains. This highlights the subjectivity of recovery and raises the question: should partial recovery be considered recovery? Or should researchers use the term “clinically significant improvement” [5]?

The PACE trial (short for “Pacing, graded activity, and cognitive behaviour therapy; a randomised evaluation”) has contributed to this debate in claiming that 22% of patients with CFS recovered after cognitive behavioral therapy (CBT), and 22% recovered after graded exercise therapy (GET) [8]. However, researchers and patients have criticized the rigor of White et al. [8] recovery criteria [9,10]. Wilshire et al. [10] reanalyzed the PACE data using more stringent criteria, as described in the original trial protocol. Under Wilshire et al. [10] analysis, CBT and GET led to recovery rates of just 7% and 4%, respectively, highlighting the need to operationalize ME and CFS recovery.

To pursue this endeavor, the ME and CFS field may benefit from examining the mental health recovery literature [11]. While physiologically-based, there are many overlapping themes in the ME and CFS field with the mental health field. Both fields lack an established biomarker. There is no blood test, or other physical measure, that determines if someone has the illness. Rather, case definitions are used to diagnose patients, although controversy exists regarding which case definition to use [12]. A second important overlap is the illness experience of patients with ME and CFS and those with mental disorders; both are chronic, invisible, stigmatized, identity-changing [13], and detrimental to individuals’ occupational, social, and leisurely lives [14]. Assessing recovery has been complicated for these chronic, invisible illnesses.

The mental health field has approached recovery using a spectrum. Davidson and Roe [15], for instance, conceptualized two types of recovery to tackle the many dimensions of recovery: “recovery from” and “recovery in.” “Recovery from” occurs when patients experience a complete amelioration of symptoms and other deficits associated with their disorder; the disorder no longer interferes with daily functioning. “Recovery in” occurs when patients still experience symptoms, but they reach a state where they can overcome the effects of being, for example, a mental patient – including poverty, loss of identity, isolation, and more [14]. Given the overlapping themes amongst chronic, physical illnesses and mental

illness, Davidson and Roe's [15] conceptions of recovery may be useful for physical illnesses that lack a definitive etiology, such as ME and CFS [11].

Because most individuals with ME and CFS have been sick for years, many may not believe a full recovery is possible, but still believe they can improve significantly [16]. Follow-up studies reflect these beliefs [17,18]. Brown et al. [17] found former patients who were deemed "recovered", defined as no longer maintaining a CFS diagnosis, still experienced significantly more impairment compared to healthy controls. Again, this raises the issue of differentiating recovery from significant improvement.

Missing from the literature is the physician perspective on ME and CFS recovery. With their diverse patient experiences, physicians could provide insight into differentiating improved and recovered individuals. However, there is variability in recovery-oriented practice among healthcare providers [19]. For instance, a patient may be treated differently depending on a providers' recovery views. Some view recovery as an outcome; others view recovery as a process [19]. Thus, there is also a need to examine physicians' recovery views to build a consensus around clinical practice. Unfortunately, there is still stigma and a lack of ME and CFS training amongst medical providers [4]. Patients often feel they know more about their illness than their providers [20]. To address this issue and fill the research gap, we explored the views of physicians with expertise in the ME and CFS field. Using semi-structured interviews, our goal was to synthesize and examine patterns among these experts regarding: (1) defining recovery from ME and CFS and (2) measuring recovery from ME and CFS.

Method

Participants

We used a non-probabilistic, purposive sample approach since physicians who treat ME and CFS are a hard-to-reach population [21], and there are relatively few experts. We tried to recruit 17 expert physicians via email. Author #4, who has worked in the field for over 25 years, provided recommendations for our list. Experts were determined by their ME and CFS patient experience, research contributions, and overall involvement in the field (e.g., running ME and CFS specialty clinics, participating on committees).

We opted for a sample of 10 individuals. This sample size was based on prior ME and CFS qualitative investigations and suggestions from Guest et al. [21], who recommend that interview structure (e.g., open vs. structured), research aims, and homogeneity influence sample decisions. The authors determined saturation when there was enough information to replicate the study and no new themes emerged with more interviews [22]. Ten experts accepted our invitation and participated in our study. Participants' average age was 65 years ($SD=12$), and eight were male. Table 1 provides descriptive information. DePaul University's Institutional Review Board provided ethics approval to conduct this research.

Procedures

Author #1 (AD) conducted semi-structured interviews with participants over the phone. Due to time, one participant completed the interview via email. Interviews were audio recorded, transcribed verbatim by AD and Author #2 (CJ), and verified for accuracy by AD and CJ.

All participants provided informed consent. Questions were adapted from our previous qualitative investigation of recovery with patients [16]. Our questions tackled physicians' thoughts on: the likelihood of recovery, defining recovery, measuring recovery, treatment approaches, and predictors of prognosis. Follow-up questions occurred naturally to delve deeper into a participant's response.

Data Analysis

We used Braun and Clarke's [23] procedure for conducting a deductive thematic analysis. Thematic analysis is a method for "identifying, analysing and reporting patterns (themes) within data" [p. 79; 23]. Using a deductive approach, a researcher codes according to their analytic interests (recovery). While a deductive approach provides a less rich account of the overall data, it allows researchers to provide a richer account of some aspect of their data, in our case, defining and measuring recovery. In qualitative research, many researchers advocate for an epistemological position to be taken as a guide for interpreting the data. Given our research aims, we applied a realist perspective – which focuses on participants' realities, meanings, and experiences – to provide a descriptive account of physicians' recovery views [24]. Concurrently, we were sensitive to latent material that is recurrent in the ME and CFS field, such as views toward etiology and stigma [25].

It is important to note the researchers' biases. AD was a research assistant with a Bachelor of Arts in psychology who had studied ME and CFS for nearly 4 years. CJ was a research intern, with a Bachelor of Arts in Ecology and Evolutionary Biology, who was newer to the ME and CFS field. Before coding, CJ immersed in the ME and CFS literature to become more sensitive to this area, particularly with recovery, qualitative methods, and controversies in the ME and CFS field. In this analysis, we felt terms like "full recovery" were redundant, since recovery marks "the act of regaining or returning toward a normal or healthy state" [26]. While "partial recovery" may be used colloquially, we feel research should not use terms like "partial" and "full recovery" because they may overstate results. Thus, we used "recovery" and "significant improvement" for our themes.

Two research questions guided our analysis: (1) How do physicians define recovery? and (2) How do they recommend measuring recovery? We coded additional features of the data to enrich our investigation (e.g., treatment approaches). Our analysis occurred in six phases. In phase 1, transcripts were uploaded to QSR's NVivo 11 software, where both coders (AD and CJ) immersed in the data through 'repeated readings' and searching for meanings and patterns. We revisited audio recordings to enhance this process. Both researchers developed their own codebook after coding three interviews (six unique interviews in total).

In phase 2, both researchers coded the same interview and met to establish a final codebook. We completed this process through comparing codes line-by-line on each segment of the interview. Barring exact phrasing, there was strong agreement of line-by-line coding and strong congruence of both codebooks. We developed a final codebook after resolving disagreements through discussion and settling phrasing [27]. The codebook included guidelines, definitions, and examples of each code. Next, we established interrater reliability on three interviews because our study was descriptive and deductive [27]. After each interview, we sought to establish moderate reliability on each individual code ($K = .70$); for

codes not meeting this threshold, we resolved disagreements, enhanced code definitions, and revisited guidelines. We calculated the weighted Kappa statistic for each code on all three interviews and the total Kappa statistic. We had good overall interrater reliability ($K=.85$) [28]. We then coded the remaining 7 interviews. AD coded 4 unique transcripts; CJ coded 3 unique transcripts.

In phase 3, we organized, summarized, and compared our data for initial theme and subtheme development. We used visual representations like mind maps and charts to enhance this process, allowing us to see relationships between themes. We refined our themes in phase 4, so each theme was distinct and maintained a coherent pattern. For this study, there was much overlap between themes. It is, for instance, impossible to discuss defining recovery without also discussing measuring recovery. Yet, we split them into two themes to display the nuances of both topics. In phase 5, we defined, finalized, and named our themes and subthemes. Author #3 and Author #4 oversaw our theme development, ensuring our analysis was credible [29]. In phase 6, we found vivid extracts to present our themes.

Results

Theme 1: Defining recovery and improvement

This theme presents a synthesis of physicians' recovery definitions. While answering the question, "What would recovery from ME and CFS look like for you?" physicians acknowledged the subjectivity of recovery. Most provided two definitions to demarcate recovery and improvement. Recovery was termed "complete recovery," "total recovery," or "full recovery." Improvement was dubbed "partial recovery" or "significant improvement." As stated, we feel it is important to delineate between recovery and significant improvement, which the following subthemes reflect.

Recovery – possible, unlikely, and subjective.—Most physicians believed only a small portion of patients recover, often citing ME and CFS literature. Physicians instead felt their patients could improve, although improvement would be more difficult for more severe patients. Participant 3, who met our expert criteria, believed recovery was common, and this was primarily because of semantics. This physician believes CFS is a temporary diagnosis; once the underlying cause is discovered, then the individual no longer has CFS.

"[L]ots of people [recover]...one of the reasons is that it has to do with semantics and labeling, that chronic fatigue syndrome is cooked from no known cause...as soon as you discover the infection, than it's no longer chronic fatigue syndrome. It's chronic fatigue from that infection, from the candidiasis, or the sinusitis."

(P3)

It should be noted this physician still provided a recovery definition for individuals suffering ME and CFS symptoms. Physician 7 did not believe CFS existed, but believed in ME, and felt the question could not be answered. Only Physician 8 believed recovery was likely in younger patients, but less likely in older patients.

“There’s always that five or seven percent who don’t recover, but my sense is that most adolescents and young adults who have chronic fatigue following some viral illness or following something else tend to recover. This is different from the women who get this disease in their forties and fifties, those probably last longer... but even those people...a lot of them recovered.”

(P8)

Overall, physicians acknowledged the subjectivity of recovery. They felt recovery breaks down to semantics and is often mislabeled. They expressed that both physicians and patients mistake significant improvement as recovery. Or, people use the phrases “partial” and “full recovery.” This is another reason why we hoped to delineate recovery from improvement.

Recovery.—Physicians viewed recovery as multidimensional, encompassing physical, psychological, and social domains. Most physicians viewed recovery as a complete remission of symptoms, where a patient returns, or is very close, to their premorbid levels of functioning. Most physicians mentioned fatigue, PEM, pain, and sleep issues when referencing symptoms.

“Recovery – to me – means going up to 12 hours a day with the caveat of being that they can push themselves and go party all night long on New Year’s Eve without having a post-exertional malaise.”

(P1)

Many physicians considered recovery as no longer meeting diagnostic criteria – though the case definitions were not always specified. However, many noted that not meeting a diagnosis provides limited information, such as quality of life. Joy and freedom, therefore, can serve as recovery indicators. Patients should be able to function in daily life confidently, without a fear of relapse. Physicians mentioned activities like showering, cleaning, cooking, and running errands. Some noted that simply watching TV, or other sedentary behaviors, would not suggest recovery. Recovered patients can exercise, take trips, and adhere to their work, social, and familial roles. They should feel confident they can achieve their goals and responsibilities, be they at work, school, or other settings.

“[I]f someone says they’re fully recovered, if they’re fully employed, if they’re leading an active social life, if they’re going to school, I think that’s pretty good.”

(P8)

Many physicians felt recovery rests on the patients – they are the ones who “define whether they have recovered” (P9). Patients know their premorbid levels of functioning and can assess what they should be able to accomplish when healthy. However, a few physicians noted this assessment becomes complicated with chronic illness, when patients are sick for many years. Thus, recovery should be assessed based on age-appropriate comparisons.

“Recovery means symptoms are no longer present, the patient is able to function at a level similar to their healthy age-level peers (I would not expect someone who became ill at 20 and recovered at age 40 to function in the same way they did at 20).”

(P6)

However, the opinions of family members and people close to the patient are also valuable, since patients may assert they are better when they are actually just improved. This confusion can arise when patients are too acclimated to their illness, so they lower their threshold and expectations for recovery. Patients may feel internal pressure to recover. Recovery may be defined as both the family and patient agreeing that the patient is recovered.

“Sometimes the patients declared themselves back to normal, but the family members shake their heads and say no he’s not, he’s better than he was, but nothing like where he was before he got sick...But, it’s the incredible statement by the patient the family that they are back to normal. That, uhh, I think is the only way to define recovery.”

(P9)

Lastly, physicians were mindful to differentiate recovery and significant improvement. Most physicians felt recovery means the patients are no longer being treated, and patients do not have to cope with their illness. Coping would indicate the patient still worries about their illness.

“‘Recovery’ to me is a strict definition and not the same as ‘mild/ moderate/ substantial improvement.’ Recovery means symptoms are no longer present... Anything short of that is merely good management of a chronic illness, which is a great goal but is not the same as recovery. For example, people with diabetes or high blood pressure may not have symptoms and function fine but they still need to take medications. Physicians do not term these patients ‘recovered.’”

(P6)

However, a few physicians, who noted the unlikelihood of a full recovery, would be happy if their patients reached a near-recovery level of functioning, contingent on some medications or coping. Nearly all of the physicians hoped to get their patients back to a level of functioning where they could perform comfortably at work or school.

“When I mentioned full recovery, I’m really talking about like the disease and all of its manifestations are gone...Our goal is to get your function to a high level that may be akin to managing Asthma, for example, where you don’t expect to cure Asthma, but you can have people on a modest number of medications that allows them to do all that you want to do, including: exercise, working, studying, that kind of thing...I would consider that pretty close to a full recovery, just acknowledging that they need some medications to support that.”

(P5)

Significant Improvement.—Physicians viewed significant improvement as noteworthy gains in functioning and a substantial reduction in symptoms, where patients may operate in daily life but still must cope or be treated. With significant improvement, the frequency and severity of symptoms are lessened. This may mean that patients experience more energy, have to sleep less, and experience periods where their symptoms are manageable.

“[Partial recovery]... that’s when you start, a person says, ‘I’m not all better, but I can, I can run twice as far now. And I only need, and I only need 8 hours of sleep instead of 10 hours of sleep.’ And, so, those, those little parameters in the history improve noticeably and consistently.”

(P3)

Depending on illness severity, some physicians considered working part-time as significant improvement, while others considered working full-time – but without the ability to perform other activities – as significant improvement.

“[It’s] them telling you that they’re able to fulfill some, but not all, of their responsibilities in the workplace if they still have them and, umm, they are somewhat, but not fully able to pursue their passions, to do the things that they want to do. Plenty of people have partially recovered from their affect.”

(P9)

Compared to recovery, patients who have improved significantly may still have to take medications or engage in other coping behaviors like pacing [30]. However, many physicians expressed they would be pleased with such progress in their patients.

“While they might be on medication directed at their various co-morbid conditions, if their function is good enough to allow them to exercise and to work fully, I would consider that pretty close to a full recovery, just acknowledging that they need some medications to support that.”

(P4)

Further, physicians felt that aiming for significant improvement was a worthy and obtainable goal, as one physician put it, “[P]artial recovery is very, umm, very possible. It’s full recovery that’s difficult” (P4). Several conveyed their patients felt hopeless, powerless, and frustrated with their illness. Reaching a significant level of functioning, however, can empower patients, alleviate uncertainty, and restore a level of normalcy.

“They’ve been sick. They are looking for answers, and umm, when they have the illness, and they know they’re seeing somebody with experience in treating it, that’s usually a relief.”

(P1)

Theme 2: Measuring Recovery

This theme presents physician’s views on measuring recovery in both practice and research. Interestingly, opinions differed on a number of measures. For instance, while some advocated using exercise tests as objective measures, others critiqued the sensitivity of such tests. However, all physicians emphasized that measuring recovery from ME and CFS should be multi-faceted. The quote below, by a research-minded physician, articulates this theme:

“Assessment of improvement or recovery should be multi-faceted incorporating different points of views (e.g. patient, clinician, family member), different symptoms (not just fatigue but PEM, cognitive function, pain, etc.), different

domains (e.g. symptoms, function), and objective (e.g. CPET, NK cell activity, neuropsychological testing, actigraphy) and subjective (e.g. SF-36, pain scales) measures.”

(P6)

Determining recovery measures is difficult. This paper presents only a few challenges associated with these suggestions.

Self-report

Everyday functioning – Activities, work, and school: Nearly all physicians emphasized general functioning as a recovery measure. Many assessed functioning in terms of hours of daily functioning – specifically upright functioning, which includes housework activities, running errands, and exercising. Some physicians, who also worked with healthy individuals, compared their ME and CFS patients to their healthy patients.

“You ask people... ‘How many hours in a day are you up and around doing something?’ And healthy adults will always say, ‘Oh, about 12–14 hours...I go to work, come home, make dinner, sit around afterwards, and we’re doing this, and we’re going to a movie.’ So about 12 hours is normal. People who have very mild ME/CFS are from about 8–12 hours; and you know, they just can’t do 12 hours.”

(P1)

One research-minded physician (P4) mentioned using the Functional Disability Inventory [31], which has patients rate items on a 1–5 scale, from “No trouble” doing said activity to “Impossible” to do said activity. Examples include “Walking to the bathroom,” “Walking upstairs,” and “Doing something with a friend.” More clinical-minded physicians also used scales, but did not mention a specific measure.

“Full recovery I would define as: the patient says he’s fully recovered. His energy levels on a scale of 1–10 are in the 8 or 9, 8 or 9 category.”

(P3)

Most physicians, either through asking patients or using a specific measure, measured functional improvement using their patient’s premorbid levels of functioning. They ask questions like: “What is [the patient] doing in the house to begin with?” (P3) “What level of functionality feels right to [the patient]?” (P9), and “What does [the patient] want to do?” (P1).

Most physicians had experience treating severely debilitated patients, who were out of work, out of school, or even bedbound. It was every physician’s goal to get their patients back in work or school. In terms of measuring recovery, however, some physicians felt work and school status were effective indicators of functioning, while others noted the limitations of such measures. On one hand, physicians viewed work and school status as positive therapeutic outcomes. Although having the ability to attend work or school does not mean a patient is no longer suffering, it shows they are somewhat able to proceed with their lives. Children may progress in school; adults can earn income, even if just part-time. Work and

school status indicate a patient has some level of functional freedom. One physician, who sees more impaired patients, said:

“[F]unctional measures – are they back in school? Are they seeing their friends? I mean...actually the biggest measure would be back in school. None of them go to school.”

(P10)

On the other hand, several viewed work and school status as “all or nothing” measures. Patients may force themselves – or in children’s cases, be forced – to attend work or school despite being severely ill. This especially occurs with patients who feel financially pressured. Further, attending work or school may be the extent of someone’s abilities, and it’s difficult to compare settings; some work requires more exertion than others. For these reasons, physicians felt researchers should use work and school measures as supplements to recovery.

“So let’s say they’re still working full-time. You say, ‘What do you do besides work,’ and they say, ‘Nothing. No I just flop down and rest and wait until tomorrow.’ Obviously, they’re not recovered very much.”

(P1)

Most problematic symptoms.—As mentioned, most physicians conceptualized recovery as complete symptom remission. This subtheme captures physicians’ views on the most impairing symptoms. All physicians were aware of the multiplicity and variation of symptoms that patients can experience. However, physicians inferred symptoms like PEM, fatigue, sleep issues, pain, and neurological issues (e.g., brain fog) to be the most problematic. Some physicians noted the importance of monitoring symptoms over time and to consider contexts like work, school, and physical activity. Assessments can occur via self-report and physicals of symptom frequency and severity.

“[W]e’ll also use what I think many people use is a 0 to 10 score, for them on frequency and their severity of different symptoms.”

(P4)

Quality of life.—Physicians felt recovery should be appraised by the patient, and, in some cases, the patients’ family. Patients may demonstrate significant improvement – reduction in symptoms, ability to work – but still feel considerably less healthy compared to their premonitory selves. Physicians noted that ME and CFS affects many facets of patients’ lives, including their identity. Patients often experience stigma from friends, family, and providers unfamiliar with ME and CFS; this experience, simultaneous with patients’ loss of abilities, increases patients’ risk of depression.

“And the doctors say, ‘Why don’t you see a shrink? Because you’re depressed.’ And I say to the patient, ‘Of course you’re depressed. Anybody who’s that sick, who’s that sick and loses 50% of the quality of their life, of course you’re depressed.’ I get depressed if I get a hang-nail.”

(P3)

Recovery, as indicated by the patient, would mean they feel free to pursue their goals and live an enjoyable life. Friends and family could help appraise this process, since patients may say, “‘Oh, I’m better. I’m recovered,’ when in fact, you really start talking to them about their symptoms and they haven’t recovered at all.”(P1)

“[W]hen I say that I believe the patient can recover, it’s because patients have told me and their friends and family have told me, ‘I am back to where I was effectively before I became ill. I am fulfilling my responses in the workplace and at home as I should and as I once did, and I am able to pursue my passions in the same way that I always did, etc.’”

(P9)

Objective Tests

Physical Functioning Measures.—Many commented on the general use of laboratory exercise measures to assess fatigue, PEM, and other symptomatology. Some supported using exercise tests in research and practice, while others mentioned their limitations. One participant (P1) specified, and advocated for, the 2 Day Exercise Test, also known as Cardiopulmonary Exercise Testing (CPET). This participant noted the test’s merits of being objective and able to measure ME and CFS severity. One suggested using actigraphy (P6) as an objective research measure; another mentioned the ability to climb flights of stairs as an exercise test.

“I ask general questions like... ‘How many flights of stairs can you climb without having to stop to rest?’ And...if they can only climb one flight of stairs, and they’re not 90 years old...then that gives me some information about, say, they’re state of conditioning.”

(P3)

Other physicians, while not rendering exercise measures useless, felt they lacked sensitivity (P5), were not the best representation of everyday functioning (P10), and were confounded by motivation (P2). Gathering other activity information – doing laundry, going to the store, cleaning – provides better daily indicators of physical functioning. One research-minded physician felt patients tend to improve cognitively, and exercise tests might exacerbate patients’ symptoms.

“I don’t really think an exercise test is really going to be the best or most sensitive way to measure progress because in my experience, when people improve, they improve cognitively, and they feel fewer symptoms, and that causes them to increase their activity, and then they have more symptoms.”

(P5)

Altogether, exercise tests may provide objective physical activity measures, especially if implemented on consecutive days. However, these tests should be supplemented with self-report measures on daily functioning. Researchers and clinicians should also note that exercise tests may flare patients’ symptoms.

Physiological measures.—Several physicians hoped for more integration of physiological measures in assessing ME and CFS. This can be difficult, however, since there is no universal, identified biomarker. Physicians provided a full medical history to rule out other manifestations.

“Well, it’s a full history, including a history with the...teenager with the parents out of the room...then a full physical examination – with the clothes off. Umm, including a detailed neurological examination.” (P10)

Some physicians examined orthostatic intolerance using the NASA 10 Minute Lean Test, which measures patients’ blood pressure upon sitting and standing [32]. One research-minded physician (P6) suggested using CPET to measure blood pressure, heart rate, and aerobic capacity, and taking blood samples to measure Natural Killer cells. Some felt future research should identify gene-expression markers in patients.

“I think we should be able to develop measures of, uhh, physiologic measures of post-exertional malaise like Gene-expression... I do believe we should be able to develop those kinds of things.”

(P5)

Most physicians were hopeful but frustrated with the state of the field. On one hand, the field has made notable progress examining the biology of ME and CFS, but there is still much progress needing to be made with developing physiological assessments.

“The brain, autonomic nervous system, the chronic activation of the immune system, the abnormalities of energy metabolism, uhh, are I think now solidly established – a long way from saying we understand continental causes of the illness – but I think it’s now pretty clear that there are underlying biological abnormalities in these patients that they are not simply imagining that they have a set of symptoms.”

(P9)

Discussion

This was the first study to examine physicians’ views on defining and measuring recovery from ME and CFS. There is a need to operationalize ME and CFS recovery. Without a consensus, treatment outcomes have been debated and overstated. Since we interviewed experts, our study provides a groundwork for how future research can approach recovery.

Like Adamowicz et al. [5], our findings highlight the importance of differentiating recovery and significant improvement. Physicians viewed recovery as complete symptom remission, a return to premorbid levels of functioning (adjusted for with age), and the ability to function in social, occupational, and physical (e.g., exercise) contexts without coping or taking medications. This recovery definition aligns with Davidson and Roe’s [15] “recovery from” concept, where an illness’s manifestations are gone and a person may operate as a healthy individual. Most physicians acknowledged this state is unlikely, as recovery rates are about 5% [6].

However, physicians were confident that patients could reach a state of significant improvement, which is a considerable reduction in symptoms, a return to moderate levels of functioning, and the ability to function in many aspects of daily life, perhaps contingent on coping strategies or medications. This definition aligns with Davidson and Roe's [15] "recovery in" concept, where an individual still has an illness, but they may overcome effects like poverty, loss of social roles, sense of self, and other consequences of being sick. This improved state fits with Fennel's [33] four-phase model of coping with CFS, which has been validated via factor analysis [34,35]. Particularly, our results fit with the Stabilization phase, where individuals integrate their pre- and post- illness self. Individuals are still sick, but they are able to function with effective illness management, similar to recovery in the mental health field. Recovery from ME and CFS may also be viewed similar to Asthma or Diabetes. Individuals still have Asthma or Diabetes, but they live fulfilling lives with illness management.

These findings align with our previous qualitative investigation with patients [16], where patients conceptualized recovery as the ability to (1) function in daily living without coping or fear of relapse, (2) returning to their identity in their many roles, and (3) be free of their most problematic symptoms. This overlap demonstrates both strength and merit for our current study. There is much literature on the divide between patients and physicians in the ME and CFS field [20,36]. That our findings align between experts and patients illustrates an opportunity to build a consensus around recovery. One difference is our physician sample viewed work status as less important relative to the patients, which is expected. Our sample comprised of many research-minded physicians, who considered confounding variables with work status.

Consistent with our patient investigation, this study suggests ME and CFS recovery should be viewed as multidimensional [16]. Recovery should be measured in terms of symptomatology, daily functioning (e.g., running errands), physical functioning (e.g., exercise), occupational functioning, and quality of life, all in comparison to patients' premorbid functioning [5]. To feasibly measure these targets, future research should build a consensus on tackling these recovery domains with specific measurements. Many studies utilize the Medical Outcomes Study 36-item Short-Form Health Survey [37], the DePaul Symptom Questionnaire [38], Chalder Fatigue Scale [39], Work and Social Adjustment Scale [40], and the Functional Disability Inventory [31]. The Functional Assessment of Cancer Therapy Scale [41] might also be adapted for ME and CFS, as the scale targets psychosocial functioning. Research should use longitudinal designs with these measures [4,5,42].

There are strengths and limitations in our study. We interviewed experts in ME and CFS, so our findings may not represent physicians who do not specialize in ME or CFS. However, that we synthesized experts' recovery views is also a strength, since our goal was to improve research and practice, and these individuals have the most patient experience. Unfortunately, this specialized group limited our sample size, and several prominent names either declined or did not have time to participate. Most of our sample viewed ME and CFS as a physiological manifestation, which may influence their recovery views. For instance, physicians who view ME and CFS as purely psychological may believe recovery is more

possible [43]. Our analysis, however, focused on defining and measuring recovery, regardless if it is possible.

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- Implications for rehabilitation
- Physicians viewed recovery as complete symptom remission and a return to premorbid functioning (adjusted for with age).
- Recovery from myalgic encephalomyelitis and chronic fatigue syndrome should be viewed as multidimensional, considering patients' daily life, psychosocial functioning, and overall physical functioning.
- These findings can improve practitioner-client interactions, as they provide recommendations for measuring recovery in research and practice.

Table 1.

Physician Characteristics

Participant	Specialty	Primary patient population	Years of experience in ME and CFS field	Interview length in minutes	*Significant research contributions
1	Pediatrics	Children and adolescents	>32	49	Yes
2	Neurology & Infectious Diseases	Adults	>28	18	Yes
3	Family Medicine	Adults	>30	48	No
4	Pediatrics	Children and adolescents	>20	30	Yes
5	General Internal Medicine	Adults	>20	30	Yes
6	Internal Medicine & Geriatric Medicine	Adults	>16	Email	Yes
7	Immunologist	Children, adolescents, and adults	>30	38	Yes
8	Pediatrics & Infectious Diseases	Children and adolescents	>20	31	Yes
9	Internal Medicine & Epidemiology	Adults	>25	35	Yes
10	Pediatrics	Children and adolescents	>20	18	Yes

* Participants with this designation have published extensively in the ME and CFS field. Many have received large-scale grants, written books, lead research clinics, and participated on advisory committees.